

## CASE REPORT

# Myositis ossificans traumatica of temporalis and medial pterygoid muscle

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**ABSTRACT**

Myositis ossificans is a rare disease that is characterized by bone deposition in the muscle or soft tissues. Myositis ossificans of the masticatory muscles is an uncommon finding. The condition is benign and results in heterotopic bone formation in the muscles of mastication, usually producing limitation of opening of the jaws. It is important to know the exact cause of the limitation of opening of the jaws for successful treatment. Computed tomographic scan and panoramic radiographs along with histological findings are essential diagnostic aids for evaluating conditions such as myositis ossificans. A rare case of myositis ossificans traumatica of temporalis and medial pterygoid muscle is presented here along with the discussion of clinical, radiological and histological features. The present case emphasizes not only on the importance of considering myositis ossificans in the differential diagnosis of limitation of opening of the jaws but also on the improvement of the overall mouth opening and treatment results.

**Key words:** Myositis ossificans, medial pterygoid muscle, trauma, temporalis muscle

**INTRODUCTION**

Myositis ossificans (MO) is a rare disease in which reactive heterotopic bone deposition occurs in muscle tissue or other soft tissues.<sup>[1]</sup> Its course may manifest itself as a hard, genetically determined, progressive systemic disease involving multiple muscle groups or be a consequence of trauma.<sup>[2]</sup> MO is divided broadly into myositis ossificans progressiva (MOP) and myositis ossificans traumatica (MOT). Myositis ossificans progressiva, also known as fibro-dysplasia ossificans progressiva, is an autosomal dominant disease in which multiple, heterotopic ossifications develop in the systemic muscle, fascia, tendons and ligaments.<sup>[3]</sup> It is associated with overexpression of the gene coding for the bone morphogenetic protein BMP4. In many cases, MOP occurs in childhood and the movement of the joints gradually becomes restricted, leading to ankylosis. In some cases, the patient may die of pulmonary complications due to restricted movement of the

respiratory muscles. The prognosis is generally poor with MOP.<sup>[2-4]</sup>

MOT is defined as a heterotrophic, non-neoplastic proliferation of bone in muscle and other soft tissues previously exposed to trauma and hematoma. It is also called as traumatic myositis ossificans, myositis ossificans circumscripta, localized myositis ossificans or fibro-dysplasia ossificans circumscripta.<sup>[3]</sup> In literature, Guy Patin in 1692 first described an extraskelatal bone formation and Von Duschlein in 1868 named it as myositis ossificans traumatica.<sup>[5]</sup> This condition has been published elaborately in the orthopedic literature suggesting of its common occurrence in the extremities. Common examples are Horse Riders Bone, Cavalrymen's Osseous Plate on the outer thigh and Infantrymen's drill bone on the deltoid. These indicate that the commonly involved muscles are quadriceps femoris, brachialis anticus, adductor muscles of the thigh and deltoid muscle. It usually appears in adolescents or young adult men.<sup>[6]</sup>

MOT is rarely found in the head or neck, including the masticatory muscles.<sup>[7]</sup> In the literature, over the past few decades, there are only a few reported cases of MOT involving muscles of mastication, out of which, two-thirds affected masseter suggesting its involvement more commonly than the rest, followed by lateral and medial pterygoid muscles. Temporalis muscle is very rarely affected and only three cases

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have been reported in the literature, where it is involved solely. Here, we report a rare case of MOT of temporalis and medial pterygoid muscles in a 21-year-old male patient along with the discussion of clinical, radiological and histological features.

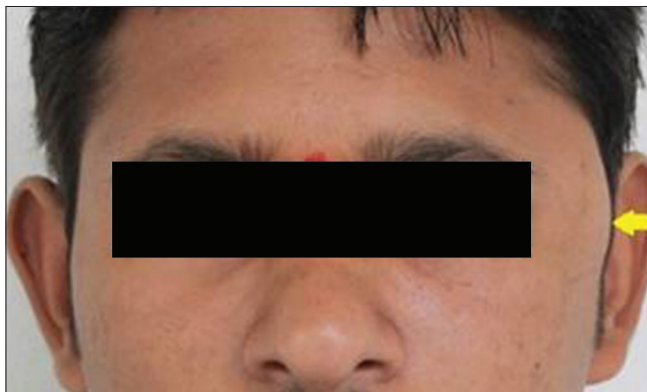
## CASE REPORT

A 21-year-old male patient reported in the Department of Oral and Maxillofacial Surgery with a progressive limitation of mouth opening and swelling on the left temporal region since two months. The patient had a history of trauma to the left temporal region two months back, which was a hit by a heavy vehicle jack rod. As there were no fractures of the facial/skull bones or any other complications at the time of injury, symptomatic treatment was given to the patient at a local hospital.

On local examination, a gross swelling was noticed in the left temporal region, which was soft in consistency and non-tender [Figure 1]. The patient had limitation of jaw movements and mouth opening was only 15 mm [Figure 2]. There were no other bony or soft tissue injuries noticed during examination. Magnetic resonance imaging (MRI) was

performed, which showed a hematoma within the temporalis muscle [Figure 3]. Thus, a diagnosis was established as hematoma and the patient was taken up for surgical evacuation of hematoma. Intraoperatively, a mouth opening of 40 mm was achieved and the patient was discharged on the sixth postoperative day with some muscle relaxants and advised forceful mouth opening physiotherapy. At the time of discharge, the patient's mouth opening was 30 mm and review on the first, third and sixth week postoperatively showed that there was recurrence of trismus, which was progressive and the patient's mouth opening was limited to 2 mm [Figure 4].

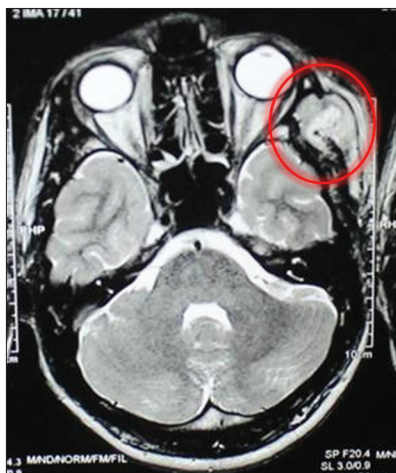
A computed tomography (CT) scan was performed, which showed calcified masses within temporalis muscle suggestive of extraskeletal bone formation [Figure 5]. A provisional diagnosis of MOT was established and the patient was taken for surgery with a treatment plan of excision of calcified masses and coronoidectomy. Intraoperatively, flakes of calcified masses were removed along with a part of temporalis muscle and ipsilateral coronoidectomy was performed. Intraoperative mouth opening of 38 mm was achieved and postoperatively forceful mouth opening physiotherapy was advised along with muscle relaxants. The excised tissue was



**Figure 1:** Clinical photograph of the 21-year-old male patient showing swelling in the left temporal region



**Figure 2:** Preoperative photograph showing limited mouth opening during the patient's first visit



**Figure 3:** MRI showing a hematoma-like lesion within the temporalis muscle



**Figure 4:** The patient's mouth opening was limited to 2 mm after first surgery in sixth week, postoperatively



sent for histopathological examination, which showed muscle fiber bundles and a classical appearance of zonal pattern with central connective tissue zone, intermediate immature bone with osteoblasts and peripheral mature bone confirming the diagnosis of MOT of temporalis and medial pterygoid muscle [Figures 6 and 7].

A planned conservative management protocol was followed and the patient was strictly advised normal physiotherapy. A six-month follow-up showed that the patient's condition had improved with an effective mouth opening of 30 mm. The present case emphasizes not only on the importance of considering MO in the differential diagnosis of limitation of opening of the jaws but also on the improvement of the overall mouth opening and treatment results.

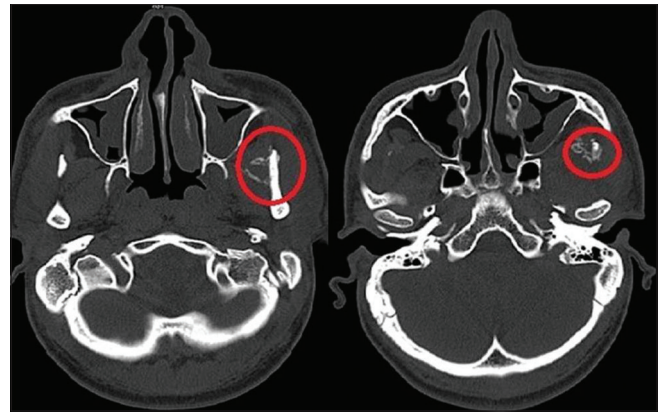
## DISCUSSION

MO, as described earlier, is heterotrophic bone formation or extraskeletal bone formation within muscle or soft tissue, including fascia and tendons. Clinically, three types of MO have been described in the literature namely congenital, traumatic and idiopathic. Congenital variant, known as myositis ossificans progressiva, also called as Munchmeyer's disease, is characterized by multiple congenital malformations and osseous metaplasia of the muscles and connective tissue, leading to progressive ossification of relevant areas.<sup>[3,8]</sup>

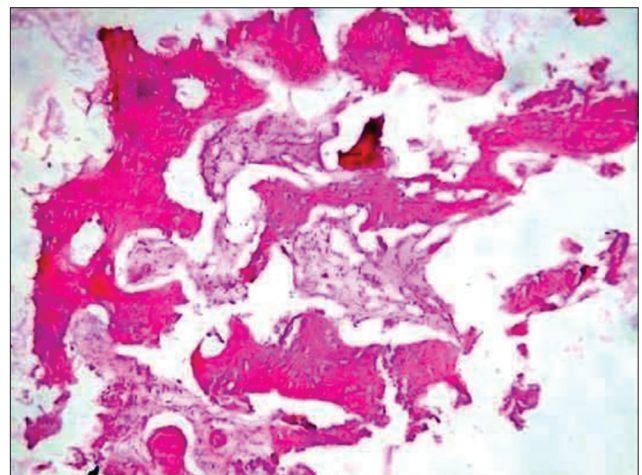
MOT is associated with a history of trauma. Depending on the area of involvement, Arrington classified MOT into stalk, periosteal and broad base muscle variants, whereas Mestan and Bassano classified MOT into parosteal, periosteal and extraosseous types.<sup>[9,10]</sup> Pathogenesis of the condition remains unclear. Carey proposed four main theories for its development: (1) Displacement of bony fragments into the soft tissues with subsequent proliferation. (2) Detachment of periosteal fragments into the surrounding tissues with proliferation of osteoprogenitor cells. (3) Migration of subperiosteal osteoprogenitor cells into surrounding soft tissues, through periosteal perforation induced by trauma. (4) Metaplasia of extraosseous cells exposed to Bone Morphogenic Proteins (BMP) derived from the lysis of bone fragments, displaced within the soft tissue during traumatic injury.<sup>[11]</sup>

The most widely accepted theory was stated by Arima *et al.* according to which, the autolysis of scattered bone fragments releases BMP that induces the differentiation of perivascular mesenchymal cells into muscular tissue resulting in a relatively homogenous bony mass. The interval between trauma and first detection of calcified mass can range from three weeks to more than 20 years.<sup>[12]</sup>

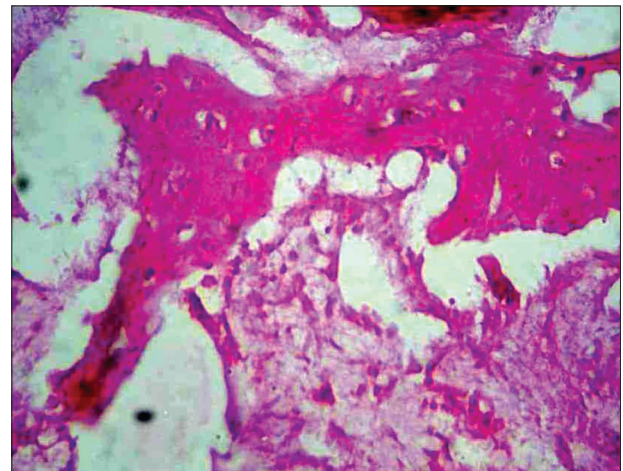
In the literature, there are about 29 cases of MOT reported in the head and neck region with male predilection and there is no age relation observed. The clinical presentation of MOT



**Figure 5:** CT showing calcified masses within medial pterygoid and temporalis muscles suggesting extraskeletal bone formation



**Figure 6:** Photomicrograph of a section of excisional biopsy specimen showing central loose connective tissue with immature bone containing osteocytes and mature bone at the periphery (H&E stain, ×100)



**Figure 7:** Photomicrograph showing central connective tissue zone surrounded by immature bone containing osteocytes in the lacunae (H&E stain, ×200)

in the head and neck region, especially when masticatory muscles are involved, is trismus. The mouth opening is limited, ranging from 1 cm to complete trismus. The limitation

of jaw movements is not sudden, instead, it is progressive. A history of trauma especially blunt in nature is evident in these presentations. The commonly involved muscle is masseter, since it is most likely to receive the force directly. Next group involved is medial pterygoid muscle followed by lateral pterygoid muscle and temporalis muscle. Though temporalis muscle is vulnerable for trauma like masseter, less chance of occurrence of MOT is seen in this group and the reason for this is unknown. In the present case report too, the patient was complaining of trismus associated with a history of trauma near the temporalis muscle.<sup>[13]</sup>

Local examination usually reveals a non-tender swelling and the affected muscles, at rest, are firm to hard in consistency. Radiographically, these lesions present themselves as abnormal calcifications within the muscle tissue. Reports suggest that it might take 3-6 weeks post injury before manifestation of symptoms and for ossification to be observed radiographically.<sup>[14]</sup> Radiographically, Shirkhoda *et al.* described four different phases of MOT: (1) The initial phase features capillary and mesenchymal cell proliferation at the periphery of the wound. Due to lack of calcification, this phase is inconspicuous on radiographic examination. (2) Initial phase of bone formation (1-2 weeks). (3) Intermediate phase (4 weeks). (4) Late phase (6 weeks). The mature phase appears as a central radiolucency surrounded by a rim of bone. Surgical excision of MOT is best performed during the mature phase of the lesion, when it is well-delineated from the surrounding skeletal muscles.<sup>[15]</sup>

Due to superimposition of cranial bones, this condition is difficult to detect on plain radiograph. In such cases, CT scan and MRI have proven to be useful; and bone scintigraphy has also been used as a modality of diagnosis. Zeanah and Hudson suggested that CT scan accurately demonstrates characteristics of well-defined peripheral shell of bone, its location and extent of the lesion into the surrounding normal tissues.<sup>[16]</sup> In the present case report too, CT scan was performed, which showed calcified masses within temporalis muscle suggesting extraskeletal bone formation through which a provisional diagnosis of MOT was established and the patient was taken for surgery with a treatment plan of excision of calcified masses and coronoidectomy.

Histologically, MOT classically presents a zonal architecture with peripheral ossification and central cellular region. Ackerman recognized and described the “zone phenomena” with inner, middle and outer zones. The central or inner zone contains undifferentiated cells, hemorrhagic and necrotic muscular tissue, loose fibrovascular tissue with spindle cells and prominent giant mesenchymal cells containing abundant mitosis. The middle zone contains active osteoblasts and immature osteoid, chondroid and woven bone tissue. The peripheral or outer zone contains mature lamellar bone with active osteoclasts and collagenous fibrous stroma.<sup>[17]</sup> Although the zonal pattern is considered the hallmark of MOT, many reports suggested that the histological

examination reveal a well-circumscribed mature osseous tissue with or without cellular component.<sup>[12]</sup> In the present case, the histopathological examination showed muscle fiber bundles and a classical appearance of zonal pattern with central connective tissue zone, intermediate immature bone with osteoblasts and peripheral mature bone confirming the diagnosis of MOT of temporalis and medial pterygoid muscle.

The differential diagnosis for MOT that should be taken into discussion can be non-neoplastic disorders such as calcified fibromatosis, phleboliths, local infections (post-traumatic periostitis, osteomyelitis) and malignant tumors such as osteosarcoma, osteochondroma and rhabdomyosarcoma. MOT, since long, was confused with osteosarcoma by many clinicians and is usually misdiagnosed as osteosarcoma because of its clinical presentation and microscopic examination. Between these two entities, there are few differences: In MOT the pain tends to decrease in time, whereas in osteosarcoma the pain tends to increase; and in MOT the calcification begins at the periphery and progresses toward the center, while in osteosarcoma it begins at the center and continues to the periphery. However, the diagnosis of MOT is based on the history of trauma, usually a blunt injury, although trauma is reported in only 70% of the cases with supportive clinical, radiographic and microscopic features.<sup>[18,19]</sup> Booth and Westers identified three important criteria to consider when diagnosing MOT, which includes: (1) A history of significant local injury; (2) Clinical and radiological evidence of ossification within two months of the initial injury and (3) The location of the lesion.<sup>[20]</sup>

Best treatment modality in MOT is prevention of further ossification and an understanding of risk factors, which may lead to formation of heterotrophic bone. Wide varieties of treatments were proposed, which include surgery and conservative management. Surgery has been an answer for MOT since long, but there had been a controversy regarding the time of surgical intervention. Many authors suggested that it is advisable to intervene only after ossification has stopped, because if surgery is attempted during immature phase there will be aggravation of ossification. If there is no improvement after conservative treatment (pharmacotherapy, physiotherapy), it is better to perform osteotomy of the coronoid process of the mandible together with the inferior attachment of the temporal muscle while leaving the calcified fibers of the temporal muscle.<sup>[21]</sup> In the present case, a planned conservative management protocol was followed, where flakes of calcified masses were removed along with a part of temporalis muscle and ipsilateral coronoidectomy was performed and the patient was strictly advised normal physiotherapy. A six-month follow-up showed that the patient's condition had improved with an effective mouth opening of 30 mm.

## CONCLUSION

It is important to know the exact cause of the limitation of opening of the jaws for successful treatment. MOT of the

masticatory muscles usually produces limitation of opening of the jaws. CT scan and panoramic radiographs along with histological findings are essential diagnostic aids for evaluating conditions such as MO. The present case emphasizes not only on the importance of considering MO in the differential diagnosis of limitation of opening of the jaws but also on the improvement of the overall mouth opening and treatment results.

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